



Surgery

A THREE-YEAR RETROSPECTIVE OBSERVATIONAL STUDY OF THE SURGICAL MANAGEMENT OF HYDATID CYSTS OF THE LIVER IN A NEWLY ESTABLISHED MEDICAL COLLEGE HOSPITAL IN SOUTH KASHMIR

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ABSTRACT

Introduction: Hydatid disease, Echinococcosis is a Zoonosis caused by cestodes of the genus *Echinococcus* (Family Taeniidae). This serious and near cosmopolitan disease continues to be a significant public health issue. It is quite common in this part of the world and majority of patients present in our Out-Patient Department with Hydatid cyst of liver who invariably need surgical intervention. We operated 47 Patients of Liver Hydatid cysts from April 2019 to March 2022 both by open (26) and laparoscopic methods (21) with majority of them undergoing endocystectomy (42) with or without omentoplasty and external tube drainage. The postoperative recovery and morbidity were shorter in those operated laparoscopically (2-4 days) as compared to open method (5-7 days) and there is no recurrence on follow up till date with insignificant postoperative complications like biliary leaks that resolved spontaneously on conservative treatment except in one patient who needed Endoscopic sphincterotomy for biliary decompression. One patient needed repair of iatrogenic diaphragmatic rent with intercostal chest tube drainage. **Aims and objectives:** To study the demographic pattern, cyst type, surgical technique, post-operative complications, and hospital stay of patients managed for Hydatid cysts of liver in the newly established Medical College Associated Hospital. **Methods:** Retrieval of the records of the patients operated electively for Hydatid disease of liver from April 2019 to March 2022 in the GMC Associated hospital Anantnag either by open or laparoscopic methods and analyzing the results for last 3 years from April 2019 to March 2022. **Conclusions:** In this study we analyzed the data of 47 patients of hepatic CE (Cystic Echinococcosis) from April 2019 to March 2022. The highest incidence of Hepatic CE was anatomically in the right lobe of liver (40) with or without synchronous cysts in other lobe or organs. The most common type of surgery performed was partial cystectomy with external tube drainage or simple endocystectomy with omentoplasty and external tube drainage (42). The most important intra-operative finding was cysto-biliary communication (7). Most of the patients had uneventful postoperative recovery. One patient needed diaphragmatic repair for iatrogenic rent due to adherence of cyst to the diaphragm and other patient needed ERCP with sphincterotomy to treat persistent significant postoperative biliary leak. The surgical technique was adopted as per operative findings like cyst size, anatomical location of cyst/cysts, number of cysts, cysto-biliary communication, cyst infection and presence of extra-hepatic Hydatid cyst or cysts. The patients were subjected to both open (26) and laparoscopic method (21) after the written consent of the patient and as per the experience of the surgeon. There is no recurrence of Hydatid CE in any of our patients in the follow up so far though the period of follow up is still shorter for recurrence (12-30 months). The patients operated laparoscopically had less morbidity and shorter hospital stay as compared to those operated by open method.

KEYWORDS : Zoonosis, Cystic Echinococcosis (CE), Alveolar Echinococcosis (AE), Peri-cystectomy, Endo-cystectomy,

INTRODUCTION

Hepatic cyst Echinococcosis (CE) is a parasitic Zoonosis caused by larval stage of *Echinococcus granulosus*. Cyst lesions occur in liver in up to 75% of the cases of CE. CE is a global parasitic Zoonosis endemic in countries in the Mediterranean Turkey, North Africa, Australia, New Zealand, South America, The Philippines, Northern China, and Indian Subcontinent (1, 2). The hepatic CE is rare in developed countries (3). The incidence in the European countries has increased because of inward migration from endemic areas (4, 5).

Hepatic CE can be treated with observational, medical, per-cutaneous, and surgical approaches.

The World Health Organization (WHO) informal working group of Echinococcus has recommended defining the treatment strategy based on the cyst stage (6). Accordingly, the patients with cysts greater than 10 centimeters or in CE stage 2 or 3b (with daughter cysts) should be treated surgically (6). However, the other cysts may also require immediate surgical treatment such as superficial cysts with a higher risk of rupture, infected cysts and cyst communicating with the biliary tree. (6)

Surgery is considered the treatment of choice for patients with large cysts and complicated hepatic CE because satisfying outcomes have been reported (7-9).

Radical surgery for hepatic CE refers to peri-cystectomy and liver resections whereas the conservative surgery involves the removal of

cyst content and sterilization of the residual cavity, also called endocystectomy together with partial cyst resection. There are several non-comparative retrospective studies concluding that radical surgery is both safe and effective (10-13).

METHODS:

We performed retrospective analysis of medical records of patients who presented to our hospital from April 2019 to March 2022. These patients presented with large single or multiple hepatic cyst who were treated by open and laparoscopic method. The following data were extracted: Age, gender, residence, Rural /Urban, symptoms and signs, radiological findings, intra-operative findings, intra and postoperative complications, duration of hospital stay and the follow up period. The diagnosis was confirmed by abdominal Ultrasonography and spiral CT scan. These radiological modalities were used to evaluate the cyst size, location, extent, and classification according to World Health Organization (WHO) (14). The data collected were entered into MS Excel sheets and analysis was carried out using statistical package for social sciences (SPSS-version 26). To explore if there is some significant difference in the proportions of different categories, we made use of Chi-Square test with $P < 0.05$ considered as statistically significant difference.

RESULTS:

47 patients were operated electively who had indication for the surgical management from April 2019 to March 2022. 32(68%) patients were females and 15(32%) were males (Table 1). Statistically

significant difference in the proportion of males and females was observed ($\chi^2_{(1,0.05)} = 6.15$; $p = 0.013$).

Most of the patients were in the age group of 20-40 years (Table 2) with ($\chi^2_{(6,0.05)} = 42.21$; $p < 0.0001$)

Most of the patients presented with abdominal pain and hepatomegaly. The number of pre-operative diagnostic tools used are shown in Table 3.

Table 1: Gender Distribution of Patients

Gender	n(%)	P-Value
Male	15(32)	0.013*
Female	32(68)	

* Distribution of disease between males and females differ significantly

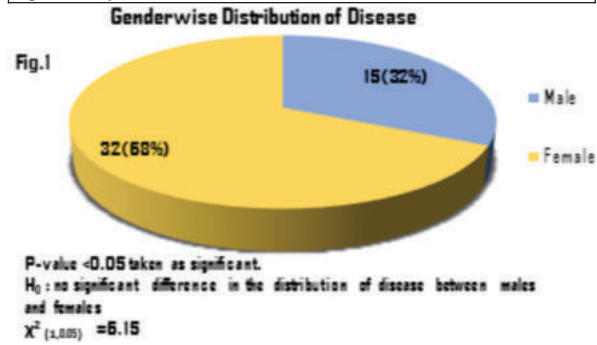


Table 2: Age Distribution of Patients

Age Group(yrs)	n(%)	P-Value
1-10	2	<0.0001*
11-20	5	
21-30	18	
31-40	15	
41-50	4	
51-60	2	
>60	1	

* distribution of disease among different age groups differs significantly

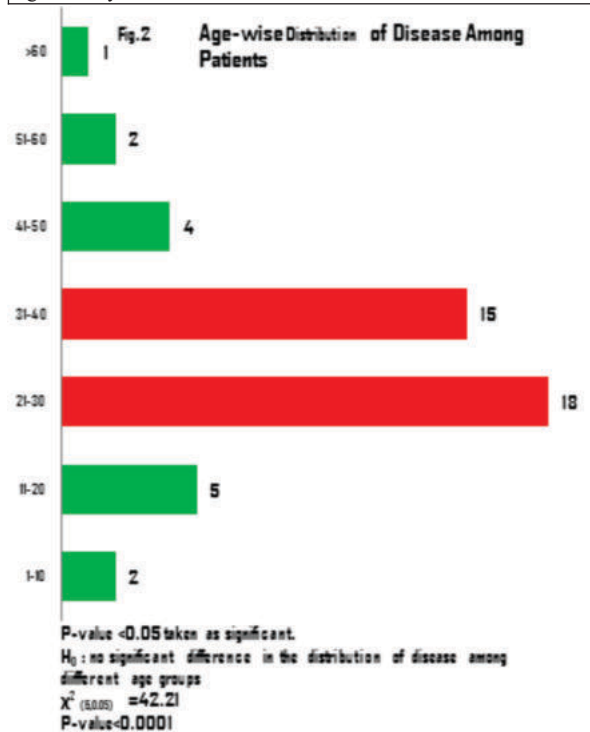
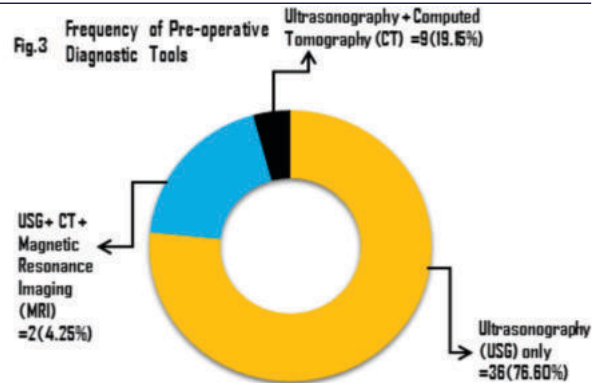


Table 3: Frequency of Pre-operative Diagnostic Tools

Diagnostic Tools	n (%)
Ultrasonography (USG) only	36 (76.60)
Ultrasonography+ Computed Tomography (CT)	09 (19.15)
USG+ CT+ Magnetic Resonance Imaging (MRI)	02 (4.25)



As per the location most of the Hydatid cysts were in the right lobe as single cyst (32), two cysts (3) or three cysts (01). There were 7 patients with single left lobe cyst (Table 4). Majority of the cysts operated belonged to Gharbi Classification Type III on Ultrasonography (Table 5). The surgical procedure done was as per the location and size of the cyst. 26 patients underwent open surgery while as 21 were operated laparoscopically.

Table 4: Site and Count of Cysts

Site	Number of Cysts		
	1	2	>2
Right Lobe	32	3	1
Left Lobe	7	0	0
Both Lobes	0	3	1

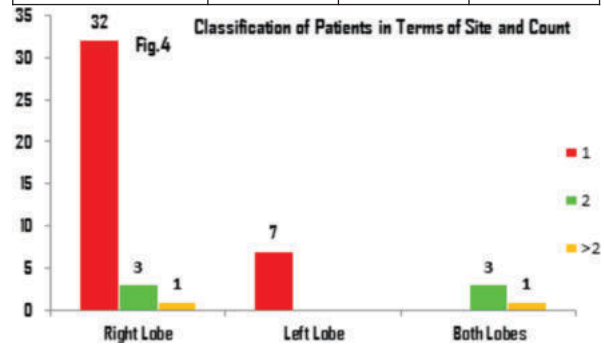
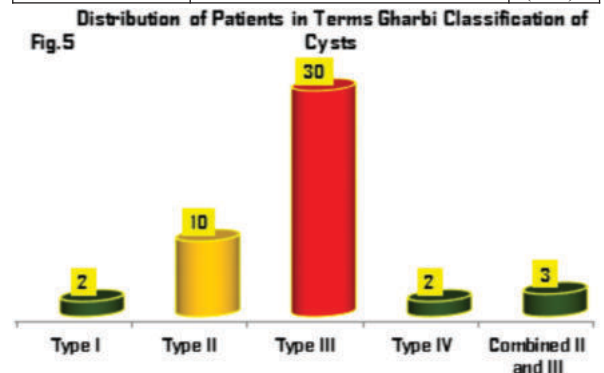


Table 5: Distribution of Patients in Terms of Gharbi Classification of Cysts

Type of Cyst	Gharbi Definition	n(%)
Type I	Pure Fluid collection	2(4.25)
Type II	Fluid collection with split wall (Water lily sign)	10(21.27)
Type III	Fluid collection with septa (Honeycomb sign)	30(63.82)
Type IV	Heterogeneous Echogenic patterns	2(4.25)
Combined II and III		3(6.38)



The procedures performed were endocystectomy (enucleation) with external tube drainage (ETD). The total surgeries conducted using this procedure being 29 out of 47, with no significant difference in the proportion of open v/s laparoscopic, ($\chi^2_{(1,0.05)} = 0.390$; $p = 0.532$). Similarly for Endocystectomy with omentopexy ($\chi^2_{(1,0.05)} = 0.160$; $p = 0.690$), Endocystectomy with capitonnage ($\chi^2_{(1,0.05)} = 0.025$; $p = 0.874$), Partial Pericystectomy with ETD* ($\chi^2_{(1,0.05)} = 0.120$; p

=0.656) and Excision of cyst (Pericystectomy) with ($\chi^2_{(1,0.05)}=0.050$; $p=0.825$). These procedures were put under practice depending on the location and size of the cyst (Table 6).

Table 6: Operative Procedures on Various Types of Surgeries

Type of Surgery	Total n=47	OPEN (n=26)	LAPARO SCOPIC (n=21)	P-Value
Endocystectomy with ETD	29(61.70)	15(57.70)	14(66.70)	0.532
Endocystectomy with omentopexy	3(6.40)	2(7.70)	1(4.80)	0.690
Endocystectomy with Cappitonnage	2(4.25)	1(3.85)	1(4.80)	0.874
Partial Pericystectomy with ETD*	8(17.02)	5(19.24)	3(14.27)	0.656
Pericystectomy (Excision of cyst)	5(10.63)	3(11.54)	2(9.52)	0.825
*ETD=External Tube Drain				
P-Value<0.05 taken as significant				
H0 : no significant difference in the percentages of two operative procedures				

The intra operative complications of diaphragmatic rent were seen in one patient and treated with the repair and intercostal chest tube drainage. Three patients underwent cholecystectomy with concomitant cholelithiasis and one patient needed cholecystectomy to get access to the cyst through gall bladder bed. The cysto-biliary leakage was found in 7 patients that was minor resulting in post-operative leak. One patient developed significant post-operative drainage of 500 to 600ml/24 hours and was treated in the department of Gastroenterology with ERCP and sphincterotomy. The leak stopped after biliary decompression. There were few insignificant post-operative complications like wound infection in 8 patients who were managed well in follow up with antibiotic therapy. The hospital stay was significantly shorter in those patients operated laparoscopically (2 to 4 days) than those operated by open method (5 to 7 days).

All the patients are under regular follow up without any significant complication or a recurrence so far.

DISCUSSION:

The presentation of Hydatid cyst of the liver depends on the localization, size, and the stage of the cyst. Although most of the Hydatid cysts of the liver are asymptomatic, the most common symptoms are the pain and hepatomegaly. Fever and jaundice may accompany complicated cases. The cysts may present as acute abdomen in case of rupture into the peritoneal cavity or biliary tract.

Ultrasonography is the primary diagnostic tool owing to its low cost, high specificity, and sensitivity. CT, MRI and MRCP may be used for better documentation and definition of the vascular/biliary anatomy. The serological methods may be useful for differential diagnosis in difficult cases (15).

High pressure intra parenchymal cysts causes communication between the cyst and the biliary system (16), resulting in jaundice, episodic cholangitis, and elevated liver function tests. ERCP and MRCP are not effective in predicting the communication of Hydatid cysts with bile ducts due to intra-cystic high pressure. However, these modalities are very sensitive in showing dilated bile ducts and daughter vesicles or germinative membranes of Hydatid cysts in bile ducts (17).

The treatment depends on stage, location, size, and complications of the cysts. Chemotherapy should be the first choice of disseminated disease and for high-risk patients for surgery. Albendazole, antiparasitic drug is recommended as the chemotherapeutic agent of choice. The usual dose is 10-15mg/kg/day (18). Franchi and colleagues treated 448 patients with Albendazole 10mg/kg/day for 6 months. The cyst degeneration was observed in 74 percent of patients while as 25percent persisted with the cyst (19). Type I AND Type II cysts can be treated successfully and effectively with percutaneous methods (20). Surgery is still the first choice for Type III, Type IV cysts and cysts opening in the biliary ducts or peritoneal cavity. There is often no need of therapy in Type V cysts (21).

The cysts located peripherally can be treated with cystectomy or hepatic resection with least possible risk of recurrence. In our study 05 patients underwent total cystectomy. However partial cystectomy with

external tube drainage was the most frequently used operation for intra-parenchymal hydatid cysts. We did omentopexy in 03 patients with the hypotheses is that it absorbs residual fluid from the cavity and stimulates macrophage migration into the operated area (22). We did capitonage technique in 02 patients believed to prevent fluid accumulation and recurrence.

Patients who have jaundice, history of cholangitis or elevated liver enzymes and dilatation or debris in major bile ducts should be assessed for main bile duct contamination (23,24). If the bile ducts are evaluated with ERCP before surgery, it is not necessary to perform main duct exploration. Kayaalp and coworkers have shown that Hydatid cysts lying in the hilar region of the liver had a higher biliary communication rate (48%) than did peripherally located cysts (27%)[25]. Alper and colleagues have shown that choledochoduodenostomy reduces mortality and morbidity rates as compared to the use of a T-tube for drainage(26).However Elbir and coworkers showed lesser morbidity and mortality, hospitalization time and infection rate with T-tube drainage(17).

An external tube drainage (ETD), is performed to prevent abscess, bilioma or biliary peritonitis. If bile drainage lasts for more than 10 days, it should be considered as fistula. ERCP may be used successfully to manage these patients with a low output, that is <100ml/day (25,26,27).

In our study 07patients had biliary leak in tube drainage that stopped in weeks' time. One patient persisted with >100ml/day drainage of bile which was managed with ERCP and sphincterotomy.

Relapse is the major problem after Hydatid surgery. Follow up Ultrasonography will not detect relapse. Result of Indirect Hemagglutination (IHA) test and an IgE radioallergosorbent test (RAST) will remain positive long after operation. The only demonstration of scolices in the remaining cavity ensures definitive diagnosis (28). The recurrence rates of the surgical techniques range between 0 and 25% but as yet no prospective randomized study has shown superiority of one operative technique over the other(29).In our study we are yet to pick up any recurrence though he follow up is still shorter to detect any recurrence.

REFERENCES:

- Buttenschoen K, Buttenschoen DC. Echinococcus granulosus infection: the challenge of surgical treatment. *Langenbeck's archives of surgery*. 2003;388(4):218–30.
- Budke CM, Casulli A, Kern P, Vuitton DA. Cystic and alveolar echinococcosis: Successes and continuing challenges. *PLoS Negl Trop Dis*. 2017;11(4):e0005477. 10.1371/journal.pntd.0005477
- Junghans T, Da Silva AM, Horton J, Chiodini PL, Brunetti E. Clinical management of cystic echinococcosis: state of the art, problems, and perspectives. *The American journal of tropical medicine and hygiene*. 2008;79(3):
- Nunnari G, Pinzone MR, Gruttadauria S, Celesia BM, Madeddu G, Malaguarnera G, et al. Hepatic echinococcosis: clinical and therapeutic aspects. *World journal of gastroenterology*. WJG. 2012;18(13):1448. 10.3748/wjg.v18.i13.1448. 301–11.
- Al-Saedi M, Khajeh E, Hoffmann K, Ghamarnejad O, Stojkovic M, Weber TF, et al. Standardized endocystectomy technique for surgical treatment of uncomplicated hepatic cystic echinococcosis. *PLoS Negl Trop Dis*. 2019; 13(6):e0007516. 10.1371/journal.pntd.0007516
- Brunetti E, Kern P, Vuitton DA. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop*. 2010;114(1):1–16. 10.1016/j.actatropica.2009.11.001
- Secchi MA, Pettinari R, Mercapide C, Bracco R, Castilla C, Cassone E, et al. Surgical management of liver hydatidosis: a multicentre series of 1412 patients. *Liver International*. 2010;30(1):85–93. 10.1111/j.1478-3231.2009.02116.x
- Safioleas M, Stamoulis I, Theohari S, Moulakakis K, Makris S, Kostakis A. Primary hydatid disease of the gallbladder: a rare clinical entity. *J Hepatobiliary Pancreat Sci*. 2004;11(5):352–6. 10.1007/s00534-004-0915-6
- Derveniz C, Delis S, Avgerinos C, Madariaga J, Milicevic M. Changing concepts in the management of liver hydatid disease. *J Gastrointest Surg*. 2005;9(6):869–77. 10.1016/j.gassur.2004.10.016
- Bildik N, Cevik A, Altintas M, Ekinci H, Canberk M, Gulmen M. Efficacy of perioperative Albendazole use according to months in hydatid cyst of the liver. *J Clin Gastroenterol*. 2007;41:312–316 [PMID:14243DOI:10.109/01.mcg000225572. 50514.e6]
- Berrada S, Essadki B, Zerouali O. [Hydatid cyst of the liver. Treatment by resection of the cyst wall. Our experience apropos of a series of 495 cases]. *Ann Chir* 1993; 47:510-512 [PMID:8215179]
- Alfieri S, Doglietto GB, Pacelli F, Costaaga G, Carriero C, Mutignani M, Liberatori M, Crucitti F. Radical surgery for liver hydatid disease: A study of 89 consecutive patients. *Hepatogastroenterology* 1997;4:496-500 [PMID:9164525]
- Alonso Casado O, Moreno Gonzalez E, Loiaz Seguro C, Calvo A, Gonzalez Pinto I, Perez Saborido B, Paseiro Crespo G, Ortiz Johansson C. Results of 22 years of experience in radical surgical treatment of hepatic hydatid cysts. *Hepatogastroenterology* 2001;48:235-243 [PMID:11268973].
- Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of the hydatid liver. *Radiology*. 1981 May;139(2):459–63. doi: 10.1148/radiology.139.2.7220891.
- Harris KM, Morris DL, Tudor R, Tohill P, Harcourt JD. Clinical and radiographic features of simple hydatid cysts of the liver. *Br J Surg*. 1986;73:835–8.
- Yalin R, Akton AO, Yegen C, Dosluoglu HH. Significance of intracystic pressure in abdominal hydatid disease. *Br J Surg*. 1992;79:1182–3.
- Elbir O, Gundogdu H, Kagitcikli M, Kayaalp C, Atalay F, Savkilioglu M, et al. Surgical treatment of intrabiliary rupture of hydatid cysts of liver: comparison of choledochoduodenostomy with T-tube drainage. *Dig Surg*. 2001;18:289–93.

18. Kern P. Echinococcus granulosus infection: Clinical presentation, medical treatment and outcome. *Langenbeck's Arch Surg.* 2003; 388:413-20.
19. Franchi C, Di Vico B, Teggi A. Long term evaluation of patients with hydatidosis treated with benzimidazole carbamates. *Clin Infect Dis.* 1999; 29:304-9. Saremi F.
20. Percutaneous drainage of hydatid cyst: use of a new cutting device to avoid leakage. *AJR Am J Roentgenol.* 1992; 158:83-5.
21. Yorganci K, Sayek I. Surgical treatment of hydatid cysts of the liver in the era of percutaneous treatment. *Am J Surg.* 2002; 184:63-9.
22. Dziri C, Paquet JC, Hay JM, Fingerhut A, Msika S, Zeitoun G, et al. Omentoplasty in the prevention of deep abdominal complications after surgery for hydatid disease of the liver: a multicenter, prospective, randomized trial. French Associations for Surgical Research. *J Am Coll Surg.* 1999; 188:281-9.
23. Sayek I, Tirmaksiz MB, Dogan R. Cystic hydatid disease: current trends in diagnosis and management. *Surg Today.* 2004; 34:987-96.
24. Vignote ML, Mino G, de la Mata M, de Dios JF, Gomez F. Endoscopic sphincterotomy in hepatic hydatid disease open to the biliary tree. *Br J Surg.* 1990; 77:30-1.
25. Kayaalp C, Bostanci B, Yol S, Akoglu M. Distribution of hydatid cysts into the liver with reference to cystobiliary communications and cavity-related complications. *Am J Surg.* 2003; 185:175-9.
26. Alper A, Ariogul O, Emre A, Uras A, Okten A. Choledochoduodenostomy for intrabiliary rupture of hydatid cysts of liver. *Br J Surg.* 1987; 74:243-5.
27. Dolay K, Akcakaya A, Soybir G, Cabioglu N, Muslumanoglu M, Igci A, et al. Endoscopic sphincterotomy in the management of postoperative biliary fistula. A complication of hepatic hydatid disease. *Surg Endosc.* 2002; 16:985-8.
28. Bozkurt B, Soran A, Karabeyoglu M, Unal B, Coskun F, Cengiz O. Follow-up problems and changes in obliteration of the residual cystic cavity after treatment for hepatic hydatidosis. *J Hepatobiliary Pancreat Surg.* 2003; 10:441-5.
29. Dawson JL, Stamatakis JD, Stringer MD, Williams R. Surgical treatment of hepatic hydatid disease. *Br J Surg.* 1988; 75:946-50.